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Case Report: An Interesting Case of a Poland Syndrome

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ABSTRACT
Poland syndrome is a rare congenital anomaly characterized by unilateral chest wall hypoplasia(pectoralis major) and ipsilateral upper limb abnormalities. This case report describes a 12 year-old female presenting with Poland syndrome associated with syndactyly of the right hand with incidental finding of Ostium secundum Atrial septal defect. The patient underwent surgical correction of the syndactyly, followed by an elective ASD DEVICE CLOSURE.

KEYWORDS: Poland syndrome, syndactyly, pediatric surgery, congenital anomalies, multidisciplinary care

I. INTRODUCTION

Poland syndrome is a rare congenital condition with an incidence of approximately 1 in 20,000 to 1 in 30,000 live births, predominantly affecting males (3:1 ratio) [1].It is generally not inherited , and no genes that contribute to the disorder have been identified.Diagnosis of Poland syndrome is based on its symptoms.Often, those with the syndrome remain undiagnosed, and some may not realize they have it until puberty.It is characterized by unilateral hypoplasia of the pectoralis major muscle, often accompanied by anomalies of the ipsilateral upper limb, such as syndactyly, brachydactyly, or hypoplasia of the hand [2]. Associated conditions may include rib abnormalities, scoliosis, and, less commonly, cardiac defects like atrial septal defects (ASDs) [3]. The etiology remains unclear, with hypothesis suggesting vascular disruptions during embryogenesis as a potential cause [4]. This case report presents a 12-year-old female with Poland syndrome, right-hand syndactyly, and with Ostium Secundum Atrial septal defect, detailing her clinical course and management.

II. CASE PRESENTATION

A 12-year-old female presented to RVM Charitable Trust Hospital, Telangana, India, with a chief complaint of fused fingers on her right hand, a condition present since birth. She was diagnosed with Poland syndrome and syndactyly of the right hand. On further evaluation was found to have an Ostium secundum Atrial septal defect. There were no other co-morbidities, and the patient had no history of allergies, previous surgeries, or significant family history of congenital anomalies.

CLINICAL EXAMINATION

Physical examination revealed a height of 126 cm, weight of 27 kg, and a BMI of 17.01 kg/m². General examination showed no pallor, icterus, cyanosis, clubbing, lymphadenopathy, or edema. Vital signs on admission were stable: blood pressure (BP) 130/70 mmHg (left arm), pulse rate (PR) 82 bpm, respiratory rate (RR) 14/min, SpO2 98% on room air. Systemic examination confirmed normal heart sounds (S1S2+), vesicular breath sounds bilaterally (BAE+), a scaphoid abdomen with no tenderness or palpable masses,with her scholastic performance below the normal limit. Examination of the right hand confirmed syndactyly affecting multiple digits, consistent with Poland syndrome-associated limb anomalies.

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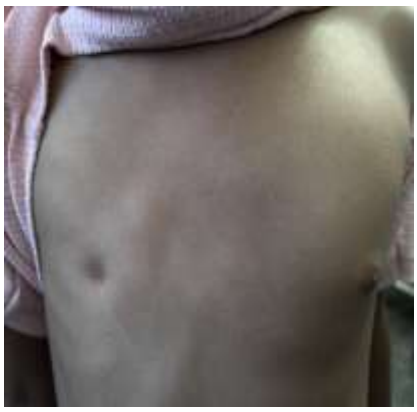
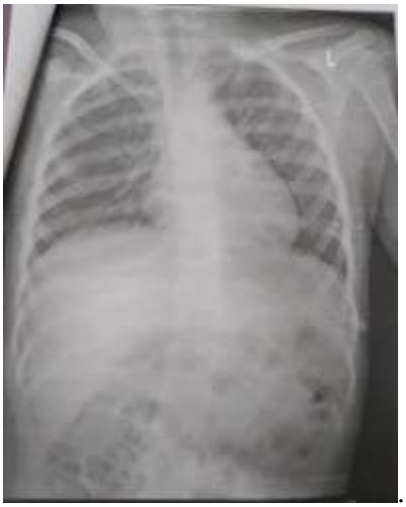
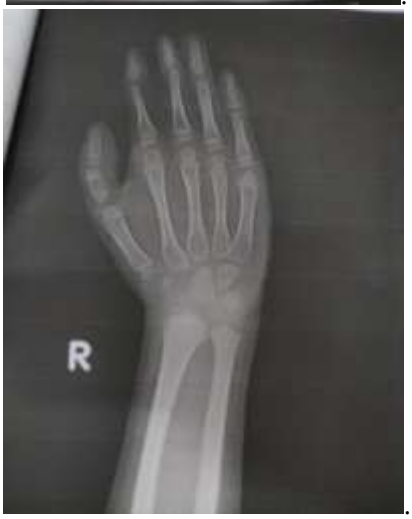
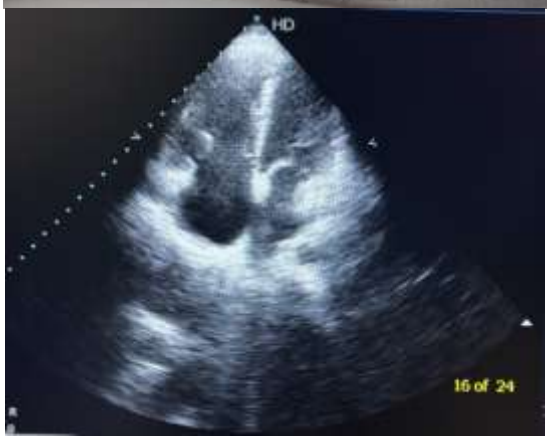


A chest X-ray revealed hypoplasia of the right pectoralis major muscle and mild rib asymmetry,with scoliosis curvature to left. An electrocardiogram (ECG) and 2Dechocardiogram were confirming the OSTIUM SECUNDUM ATRIAL SEPTAL DEFECT of 7-8 mm , with qp/qs ratio 2:1,with mild dilated RA,RV , No PAH,No LSVC,Normal mitral valve .







DIAGNOSTIC INVESTIGATIONS

Laboratory investigations included a biochemical profile, hematology report, and serology:
Hemoglobin (Hb): 11.9 g/dL (reference range: 11–13 g/dL)
White blood cell count (WBC): 9700 /cu.mm (reference range: 4000–11,000 /cu.mm)
Platelet count: 2.53 lakhs/cu.mm (reference range: 1.5–4.5 lakhs/cu.mm)
Serum sodium: 144 mmol/L (reference range: 135–145 mmol/L)
Serum potassium: 4.60 mmol/L (reference range: 3.5–5.1 mmol/L)
Blood urea: 18 mg/dL (reference range: 10–43 mg/dL)
Serum creatinine: 0.4 mg/dL (reference range: 0.3–0.7 mg/dL)
International normalized ratio (INR): 1.32 (reference range: 0.8–1.2)
Serology for HIV, HCV, and HBsAg: Negative

The elevated INR prompted further evaluation, but no bleeding tendencies were noted clinically. A surgical profile confirmed fitness for surgery after pediatric and cardiology consultations

III. MANAGEMENT

The patient underwent surgical correction of the right-hand syndactyly under general anesthesia,involving contracture release and digit separation.Vitals remained stable throughout the hospital stay.The team ensured regular vital

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monitoring, medication administration, and pain assessments, with the patient remaining stable.

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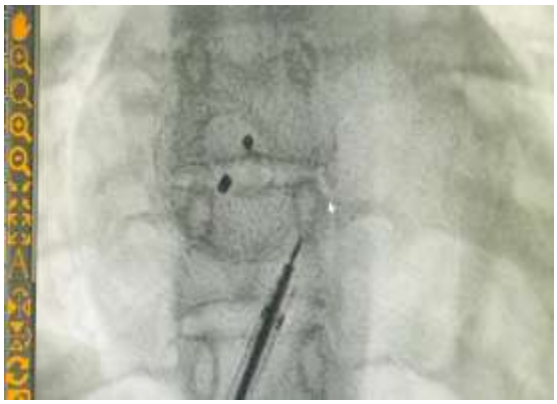
The patient's postoperative course was uneventful, with a stable clinical condition report. Following after went for an ELECTIVE ASD DEVICE CLOSURE FOR 7-8 mm OSTIUM SECUNDUM ATRIAL SEPTAL DEFECT with LIFETECH CERAFLEX ASD OCCLUDER DEVICE.



IV. DISCUSSION

Poland syndrome presents a spectrum of anomalies, with the hallmark being unilateral pectoralis major hypoplasia and variable upper limb defects [5]. Syndactyly, as seen in this patient, is a common associated anomaly, reported in approximately 10–15% of Poland syndrome cases [6]. The presence of an ostium secundum atrial septal defect in this patient aligns with rare reports of cardiac anomalies in Poland syndrome, though the exact association remains unclear [3]. Surgical correction of syndactyly in such cases aims to improve hand function and aesthetics, with studies showing favorable outcomes in pediatric populations when performed before school age [7].Preoperative evaluation, including hematology and cardiology assessments, ensured surgical safety, particularly given the history of ASD. The elevated INR (1.32) warranted monitoring, though no clinical bleeding was observed, consistent with literature suggesting that mild INR elevations in children may not always necessitate intervention unless symptomatic [8].Postoperative care focused on pain management, infection prevention, and monitoring for complications. The use of prophylactic antibiotics and anti-inflammatory agents aligns with standard protocols to reduce surgical site infections and edema [9]. The patient's stable vitals and low pain scores indicate a successful early recovery, though long-term follow-up is essential to assess functional outcomes [10].

V. CONCLUSION

This case highlights the successful management of Poland syndrome with syndactyly with Ostium secundum Atrial septal defect in a 12-year-old female, emphasizing the importance of a multidisciplinary approach. Early surgical intervention, comprehensive preoperative evaluation, and meticulous postoperative care contributed to a favorable outcome. Clinicians should be aware of potential associated anomalies, such as cardiac defects, and tailor management accordingly. Future research should focus on the long-term functional outcomes of syndactyly repair in Poland syndrome and the underlying mechanisms of associated cardiac anomalies like OS ASD.

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